

Gastric adenomyoma determined incidentally during sleeve gastrectomy: A case report

Gastric adenomyoma

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Abstract

Gastric adenomyoma is a rare benign tumor, characterized by gland-like structures embedded within a smooth muscle stroma. We report the case of a 26-year-old woman with a body mass index (BMI) of 51 kg/m2 who was not having any metabolic diseases was prepared for obesity surgery. No pathologies were detected in the abdominal ultrasound or upper gastrointestinal system endoscopy taken before the operation. Abdominal computerized tomography was obtained which revealed a nodular lesion causing mural thickening on the anterior wall of the antrum. The mini-gastric bypass surgery procedure was performed with subtotal gastrectomy to remove the mass. The final diagnosis of gastric adenomyoma was made. Although gastric adenomyomas are rare, they should always be kept in mind in the differential diagnosis of extramucosal gastric lesions.

Keywords

Adenomyoma; Gastrectomy; Computerized Tomography

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Introduction

Gastric adenomyoma has a reported incidence of 0.6 to 5.6% with the majority being found in the upper gastrointestinal tract within 5 to 6 cm of the pylorus, many within the stomach itself [1]. The tumor is produced from the glandular structures embedded in the smooth muscle bundles some of which are lined by cystically enlarged columnar epithelium [2]. This tumor is first defined by Magnus-Alslebenn in the year 1903 [3]. The patients with gastric adenomyoma may be asymptomatic, or they may show non-specific symptoms such as epigastric pain or vomiting [4]. Herein we will report a patient incidentally diagnosed with adenomyoma during sleeve gastrectomy.

Case Report

A 26-year-old woman with a body mass index (BMI) of 51 kg/ m2 who was not having any metabolic diseases was prepared for obesity surgery. No pathologies were detected in the abdominal ultrasound or upper gastrointestinal system endoscopy taken before the operation. The preparation of the operation was completed, and laparoscopic sleeve gastrectomy procedure was planned. During the operation, a lesion about 2 cm in diameter was determined in the gastric pre-pyloric region (Figure 1). The planned surgical procedure was not performed because the mass was very close to the pyloric ring. Abdominal computerized tomography (CT) was obtained which revealed a nodular lesion causing mural thickening on the anterior wall of the antrum (Figure 2). T2-weighted axial magnetic resonance imaging (MRI) with fat suppression revealed the hypointense mural nodule with the underlying normal mucosa (Figure 3). This hypointensity is thought to result from the high mixoid component. In this MRI study that was previously performed for other reasons, no contrast-enhanced images were obtained. Written informed consent was obtained from the patient who participated in this case. The mini-gastric bypass surgery procedure was performed with subtotal gastrectomy to remove the mass. In the macroscopic evaluation, there was an intramural gray-white colored mass lesion observed in 1.6x1.5 cm dimensions with irregular borders causing a protrusion in the serosal surface in the antrum. In microscopic investigations, there were glandular structures under the gastric mucosa and pancreatic acinar glands in muscularis propria between hypertrophied muscle bundles lined with columnar and flattened mucinous epithelium some of which were cysticly enlarged (Figure 4, 5). There was no atypia or mitotic activity in the tumor. Immunohistochemical

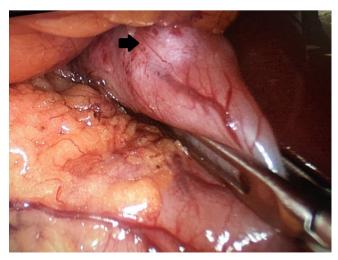


Figure 1. Macroscopic showing nodular mass in the stomach serosa (black arrow)



Figure 2. Axial CT with oral and IV contrast material shows nodular mural thickening of anterior antral wall of stomach (arrows). Note that the perigastric fat planes are fine without any finding of invasion.

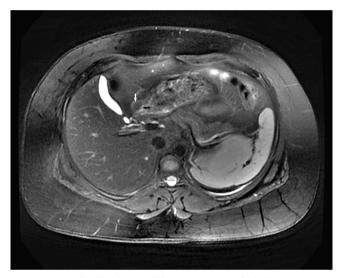


Figure 3. T2-weighted axial MRI with fat suppresion reveals the hypointense mural nodule (white arrow) with the underlying normal mucosa (black arrow).

staining revealed positive staining in smooth muscle bundles with smooth muscle actin (SMA). Stromal component revealed low proliferative index of Ki-67 (1%) and p53 less than 1% in both epithelial and stromal components. CK7 revealed positive staining in epithelial component. With all those findings, the patient was diagnosed with gastric adenomyoma.

Discussion

Adenomyoma is a lesion characterized by epithelial and smooth muscle proliferation. The epithelial origin of the adenomas is unidentified. After birth, undifferentiated glandular structures were suggested to be developed from the embryonic epithelial buds that were remaining in mature pancreatic and duodenal tissues. On the other hand, smooth muscle tissue, differing from the epithelial tissue, was suggested to be developed from the embryonic muscle bud or as a result of the stimulation of normal muscle tissue by mislocalized epithelium [5, 6]. Some authors also suggested that; adenomyoma should be regarded as an incomplete pancreatic heterotrophy due to the incorrect localization of pancreatic exocrine and endocrine components [7]. On the other hand, Takeyema et al. suggested the idea of a hamartomatous lesion that takes place due to a dysembryonic

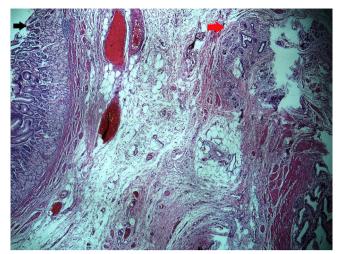


Figure 4. Microscopy showing normal gastric mucosa (black arrow) and fibromuscular stroma below the submucosal layer with a branched gland (red arrow) (HE, x 40)

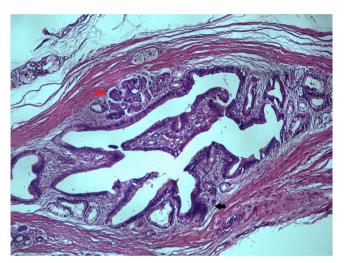


Figure 5. Microscopy showing glands lined by columnar mucinous epithelium (black arrow) and pancreatic acinar glands (red arrow) (HE, x 200)

mechanism [8]. For that reason, in its naming, some different terms such as myoglandular hamartoma, myoepithelial hamartoma, adenomyomatous hamartoma, and adenomyosis are in use [9].

The mean diameter of the adenomyomas range between 0.6-4.5 cm, and generally, they are located in submucosal or sometimes muscular layers. This disease may be seen in patients aged between 8 months to 82 years which is more common in males than females. Although adenomyomas may be reported in all parts of the gastrointestinal tract; they are most commonly determined in the fundus of the gallbladder and gastric antrum or pylorus [5].

Although in preoperative diagnosis, CT and endoscopic ultrasound are in use, it is still difficult to diagnose gastric adenomyomas in the preoperative period [2]. With endoscopic examination, it is not possible to differentiate gastric adenomyomas from other extramucosal gastrointestinal tumors, lipoma, neurilemma, hemangioma, carcinoid tumor, lymphoma, or gastric carcinomas. In many of the cases, an endoscopic biopsy is obtained, but this procedure is superficial, and unsuccessful to manage the tumor. For that reason, in intraoperative diagnosis, frozen is very beneficial in order to prevent unnecessary large operations.In histological evaluations, it is important to differentiate gastric adenomyoma from the high-grade adenocarcinoma or gastritis cystica profunda. There are some gastric

adenocarcinomas reported to be associated with the gastric adenomyomas [10]. Adenocarcinomas are differentiated from the adenomyomas with more commonly determined epithelial atypia, mitotic figures or fibrous stromal reactions. On the other hand, gastritis cystica profunda is differentiated from gastric adenomyoma with its characteristic finding of cystic, dilated gastric glands lying through the gastric submucosal layer and elongation of hyperplasic foveolar epithelium and absence of smooth muscle buds around the cyst.In the treatment of gastric adenomyomas, wide surgical excision or endoscopic submucosal dissection is enough, but surgical procedures may differ regarding the localization [2].

In conclusion; although gastric adenomyomas are rare, they should always be kept in mind in the differential diagnosis of extramucosal gastric lesions. In suspected cases without the preoperative diagnosis; we believe that frozen would be beneficial in the prevention of unnecessary radical operations.

Scientific Responsibility Statement

The authors declare that they are responsible for the article's scientific content including study design, data collection, analysis and interpretation, writing, some of the main line, or all of the preparation and scientific review of the contents and approval of the final version of the article.

Animal and human rights statement

All procedures performed in this study were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards. No animal or human studies were carried out by the authors for this article.

Conflict of interest

None of the authors received any type of financial support that could be considered potential conflict of interest regarding the manuscript or its submission.

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